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Case Report

Avascular necrosis of right mandible secondary to takayasu arteritis - A case report

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ABSTRACT:
Avascular necrosis (AVN) of the mandible is an extremely rare condition, often associated with systemic diseases or trauma, but its occurrence secondary to Takayasu arteritis (TA) is particularly uncommon. Takayasu arteritis, also known as "pulseless disease," is a chronic, systemic vasculitis characterised by inflammation and subsequent damage to the medium- and large arteries and their major branchesleading to arterial stenosis, occlusion, and reduced blood flow.We report a 50-year-old female presented with pain concerning the right lower premolar area, which was not relieved by root canal treatment. Radiographic and CBCT imaging revealed features consistent with osteomyelitis, including apical radiolucency and lingual cortical disruption. Due to compromised vascular supply and high anaesthesia risk, a careful intraoral sequestrectomy was performed with primary closure. The patient showed satisfactory recovery, with wound dehiscence managed conservatively. At one-year follow-up, the patient was symptom-free, with no signs of recurrent osteomyelitis.
Keywords: Takayasu Arteritis Osteomyelitis Pulseless Disease

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INTRODUCTION

Avascular necrosis (AVN) of the mandible is an extremely rare condition, often associated with systemic diseases or trauma, but its occurrence secondary to Takayasu arteritis (TA) is particularly uncommon(1). Takayasu arteritis is a rare idiopathic systemic chronic inflammatory arteritis that predominantly impacts the aorta and its main branches. Commonly known as pulseless disease, it is also referred to as aortic arch syndrome, young female arteritis, idiopathic aortitis, reversed coarctation, or Martorell syndrome(2). This condition has a global annual incidence of approximately 2.6 cases per million people, with women being more commonly affected than men(3). The etiopathogenesis of Takayasu arteritis (TA) remains undefined, with infections, autoimmune mechanisms, and genetic predispositions being key areas of investigation(3). The ischemic sequelae of TA can extend to various regions of the body, but the involvement of the mandible is rarely documented(4). In patients with systemic conditions such as Takayasu arteritis, osteomyelitis management becomes challenging due to compromised vascularity and the need for careful consideration of immunosuppressive therapy(5). This case highlights the surgical management of mandibular osteomyelitis in a patient with Takayasu arteritis, including considerations for preoperative risk assessment, surgical approach, and postoperative care.

CASE REPORT

A 50-year-old femalewith a known history of Takayasu arteritis (type 1) presented with persistent pain in the right lower premolar area, which remained unrelieved following root canal treatment. On clinical examination, a draining sinus with pus discharge was noted periapically. Her medical history included four years of corticosteroid and antiplatelet therapy, with absent pulses in both upper extremities. A dental pantomogram (OPG) revealed an irregular radiolucency extending from the apices of the first and second premolars. Cone-beam computed tomography (CBCT) further suggested osteomyelitis, with evidence of a fracture in the lingual cortex of the

right mandible.A biopsy revealed yellowish necrotic bone without granuloma or neoplastic features. Despite initial efforts to manage the infection, the biopsy site failed to heal, and continuous pus discharge with a fetid odour persisted (fig 1,2,3).

Preoperative Assessment

Due to the patient's complex medical history and the potential risks associated with general anaesthesia, a preoperative assessment was conducted. Carotid Doppler ultrasonography revealed a narrowing of the bilateral common carotid arteries (60-70%), indicating a high risk of stroke and other perioperative complications. High-risk consent was obtained from the patient's relatives, and the surgical procedure was planned to minimise operating time and reduce potential complications related to anaesthesia and compromised vascularity.

Since the patient had diminished blood supply to the head and neck, an intraoral approach without any reconstruction was selected to avoid potential complications from an extraoral approach and the poor prognosis of flaps or grafts. A ridge incision was made to access the necrotic bone. The yellowish-brown necrotic tissue was identified and sequestrated (fig 4) and buccal decortication was done. The surgical site was closed primarily, and sharp bony edges were trimmed to prevent soft tissue necrosis. A Soframycin-Metrogyl-impregnated gauze pack was placed to prevent dead space formation.

Postoperative Management and Outcome

The gauze pack was removed on the third postoperative day, and soft tissue closure was intact without dehiscence for the first week. However, dehiscence of the wound occurred in the later follow-up, which was managed conservatively. At four weeks postoperatively, a well-epithelialized bone cavity was observed at the site of dehiscence (fig 5). At one-year follow-up, the patient remained asymptomatic, with no clinical or radiological evidence of osteomyelitis recurrence.

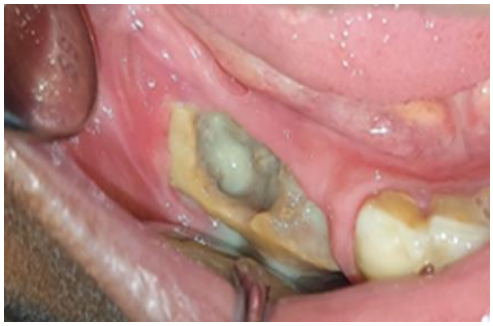

Fig: 1 Exposed necrotic bone

Fig 2: OPG showing mixed radiolucency and radioopacity41 to 47 region

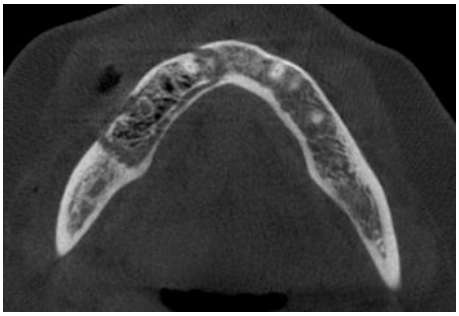

Fig 3: CBCT showing sequestrum

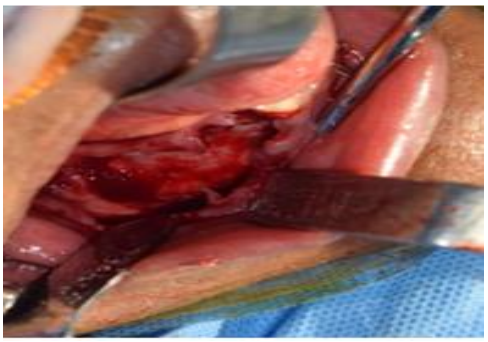

Fig: 4 After sequestrectomy and buccal decortication

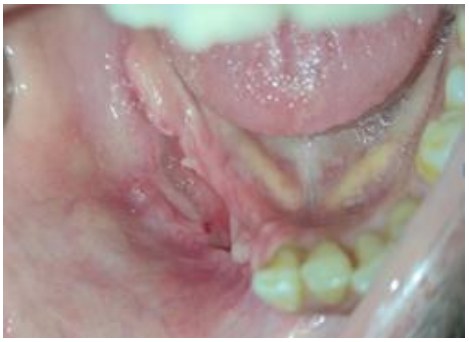

Fig: 5Postoperative healing of the surgical site

DISCUSSION

Osteomyelitis of the mandible secondary to Takayasu arteritis (TA) represents a rare but significant clinical entity, highlighting the interplay between systemic vascular pathology and localised mandibular disease(6). This case contributes to the limited literature by describing a unique presentation ofosteomyelitis secondary to TA, managed successfully with a multidisciplinary approach.

Most reported cases involve non-craniofacial bones, such as long bones and vertebrae, reflecting the systemic nature of TA-induced ischemia. Mandibular involvement, as seen in the present case, has only been reported in two previous instances, where primary chronic osteomyelitis of the mandible coexisted with TA in a younger patient. Unlike those cases, which involved younger females, the present case involved a 50-year-old patient, highlighting a broader age range for mandibular presentations in TA(7,8).A recent case report by Shirai et al(7). highlighted the rarity of chronic osteomyelitis in patients with Takayasu arteritis (TA), identifying only six prior cases in the literature and estimating its prevalence among adult TA patients at 1.47%.Our patient exhibited a persistent non-healing sinus in the mandibular premolar region despite root canal treatment, paralleling the challenging presentations often seen in TAK-associated osteomyelitis. Advanced imaging revealed significant pathological changes, including a fracture in the lingual cortex, which is indicative of the severity of the bone involvement. In contrast to the younger age profile described in the majority of the previously reported cases, our patient represents a rare occurrence in an older individual, broadening the clinical spectrum of TA-related mandibular osteomyelitis. Both cases and the one presented by Shirai et al. highlight the impact of TA-related vascular insufficiency as a significant factor contributing to the condition's poor healing response and chronic nature. However, while Shirai et al.'s report emphasised sterile inflammation as the likely underlying mechanism, our patient demonstrated overt signs of infection with persistent pus discharge, a fetid odour, and necrotic bone on biopsy. This suggests that while the vasculitis associated with TAK creates a predisposition to osteomyelitis through ischemia and impaired bone healing, superimposed infection can significantly worsen the clinical course.In both cases, surgical intervention was crucial for the removal of necrotic tissue, but the approaches differed slightly due to patient-specific factors. In our case, an intraoral approach was chosen to avoid complications related to the patient's high surgical risk due to severe carotid artery stenosis and impaired vascularity. The careful preoperative planning and use of conservative postoperative management resulted in favourable long-term outcomes, with no recurrence of osteomyelitis after one year.Unlike Shirai et al.'s patient, our case underscores the challenges posed by advanced age, longstanding corticosteroid therapy, and superimposed infection. These factors likely contributed to delayed healing and the wound dehiscence observed postoperatively, necessitating conservative secondary management. The importance of a tailored surgical and medical approach in such complex cases cannot be overstated. Døving et al(8).'s 27-year-old patient with TA had cortical thickening and signs of chronic inflammation in the mandible, with less severe vascular involvement, and underwent buccal decortication with observed trabecular bone loss. While both cases highlight the challenges of treating osteomyelitis in TA due to reduced vascularity, our patient's more advanced vascular compromise necessitated a more conservative surgical approach, resulting in a favourable outcome with complete resolution of the infection at one-year follow-up.In terms of medical management, most reported cases of osteomyelitis in TA have responded to corticosteroid therapy. However, in this patient, who was already on steroid therapy, further medical management options were limited, and surgical intervention became the primary approach to manage osteomyelitis.

CONCLUSION

Overall, this case further illustrates the complexity of osteomyelitis in TAK patients, emphasising the multifactorial nature of the condition. It highlights the need for early diagnosis, multidisciplinary planning, and individualised management strategies to address both the vascular and infectious components of the disease.

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Conflict of Interest

The authors declare no conflict of interest related to the content of this case report. The study was conducted independently, and no financial support was received from any external organisations or commercial entities.

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